MINI REVIEW

Pathways to causation and surgical cure of chronic pelvic pain of unknown origin, bladder and bowel dysfunction – an anatomical analysis

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Submitted: Oct. 14, 2018 Accepted: Dec. 12, 2018 Published online: **Introduction** Current thinking is that chronic pelvic pain of unknown origin (CPPU) is poorly understood and its treatment is empirical and ineffective. According to the Integral Theory System (ITS), however, CPPU is secondary to uterosacral ligament (USL) laxity which is associated with bladder and bowel symptoms and all are potentially curable by surgical reinforcement of USLs.

Material and methods We applied the ITS to anatomically explain the pathogenesis and cure of these conditions.

Results The first mention of CPPU being caused by lax USLs was in the pre- WWII German literature by Heinrich Martius. CPPU was first described in the English literature in 1993 as one of the four pillars of the Posterior Fornix Syndrome 'PFS' (CPPU, urgency, nocturia and abnormal bladder emptying). Surgical cure/improvement of CPPU was achieved by shortening and reinforcing USLs initially with USL ligament plication and later with tensioned tapes because of deteriorating cure rates. Non-invasive 'simulated operations' which support USLs in the posterior fornix help predict USL causation.

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Conclusions USL tapes cure/improve CPPU, bladder and bowel dysfunctions by reinforcing the USLs against which the 3 directional forces contract. Weak suspensory ligaments may invalidate these forces to cause incontinence, emptying and pain symptoms, all of which can be potentially reversed by using tapes to reinforce the damaged ligaments, as demonstrated.

Key Words: bladder emptying \diamond chronic pelvic pain of unknown origin \diamond fecal incontinence \diamond integral theory \diamond overactive bladder \diamond posterior fornix syndrome \diamond uterosacral ligaments

INTRODUCTION

Chronic pelvic pain of unknown origin (CPPU) in the female is a mystery to the studied bodies. Learned institutions such as the International Continence Society (ICS) recognize that CPPU is associated with bowel and bladder dysfunctions, but do not know its origins and do not consider CPPU to be curable [1]. The findings of the 2005 Cochrane Review were similar to those of the ICS: chronic pelvic pain (CPPU) was common in women; it caused disability and distress; laparoscopy revealed no obvious cause for the pain; pathophysiology was not well understood; treatment of CPPU was often unsatisfactory and limited to symptom relief; the main approaches to treatment include counseling or psychotherapy [2]; depression, and psychological disturbance being considered causative of CPPU [1, 2, 3], possibly a consequence of Beard's studies in the 1980's which showed a strong correlation between the two [3]. Yet simple logic dictates that depression and psychological disturbance could equally be a consequence of the severe pain [4], as evidenced by the reports of many investigators in the field, who found that cure of CPPU and of overactive bladder (OAB) symptoms, when they did occur, occurred immediately, the day after the operation [5–14] and psychological disturbances lifted at the same time [4].

We analyzed previous reports of CPPU cure [5-14] with particular focus on a large series of 611 patients who were cured of CPPU (n = 194), urgency (n = 317), frequency (n = 310), nocturia (n = 254) and fecal incontinence (n = 93) by surgical reconstruction of the cardinal and uterosacral ligaments [5], Figure 1, as predicted by the Integral Theory [15, 16]. One aim of this work was to shed light on the mechanisms for pathogenesis and symptom cure of CPPU and its associated bladder and bowel symptoms as reported [5]. Another aim is to help relieve the incredulity expressed by many colleagues that surgical cure of such widely variable symptoms is achievable by a simple uterosacral ligament (USL) sling.

MATERIAL AND METHODS

We reviewed data [5–14] of surgery based on the Integral Theory System (ITS), the most recent iteration being in 2018 [16].

RESULTS

It was found [5–17], that the pain, bladder and bowel symptoms occur in predictable groupings. This allows their ligamentous origin to be accurately diagnosed with reference to the pictorial diagnostic algorithm. Figure 1 [16]. The first description of CPPU being caused by uterosacral ligament laxity in the English literature was in 1993, as part of the 'posterior fornix syndrome' (PFS) [15]. PFS was initially comprised of CPPU, urgency, nocturia and abnormal bladder emptying, but was later expanded by Abendstein to include anorectal conditions. These symptoms were cured or improved by USL repair [5-17]. However, Goeschen [17], pointed out, that USL as a cause of CPPU was described in the German literature by Heinrich Martius in 1938. Martius stated that in about 30% of cases, backaches are provoked by damaged suspending or supporting ligaments of the pelvic organs. The paired 'ligamenta rectouterina', which are connected via paraproctium to the bony sacrum are termed 'plica or ligamenta sacrouterina'.

The causation pathway for chronic pelvic pain of unknown origin is referenced in Figure 1; the sacral (S2–4) and Frankenhauser (T11–L2) plexuses are sited at the distal ends of the USLs. It was hypothesized [18], that collagen defect in the USLs made them unable to structurally support the nerve plexuses as shown in Figure 1, so that the force of gravity pulling on them stimulated them to cause referred pain. This explains how a large majority of such patients state that the pain worsens during the day and is relieved on lying down [18]. The pain is referred in the following forms and sites:

- persistent abdominal pain in the lower part of the body, often unilateral (T11–L2);
- lower sacral pain (S2-4);
- paraurethral pain and tenderness (S2–4);
- introital, perineal pain (vulvodynia) (S2-4);
- dyspareunia (S2–4) and post-coital pain (T11–L2);
- possibly the bladder pain of interstitial cystitis.

The constant pain may cause asthenia, irritability and depression. It is the experience of surgeons who follow the ITS, that cure of these pains, when they occur, is virtually instantaneous.

Causation pathway for urge, frequency, nocturia which co-occur with chronic pelvic pain of unknown origin

One of the main discoveries from the prototype midurethral sling (MUS) operations in 1990 was that three opposite directional striated muscle vectors act around the pubourethral ligament (PUL) to stretch the anterior vaginal wall in the manner of a trampoline, to support the bladder base stretch receptors 'N', Figure 1.

Simplistically, the forward vector pulls the vagina forwards against PUL. The backward vectors pull back and down against USL, Figure 1. According to experimental data detailed in the 1990, 1993 and 2018 iterations of the Integral Theory: urge incontinence, frequency, nocturia are different expressions of a prematurely activated, but otherwise normal micturition reflex [16], caused by inability of the vagina to support the stretch receptors 'N', Figure 1. These fire off at a lower bladder volume to activate the micturition reflex. The afferent impulses are interpreted by the cortex as 'urgency' and at night 'nocturia'. Because urine storage capacity is impaired, the patient urinates more frequently. Any looseness in PUL or USL will inhibit the ability of the opposite muscle vectors to support 'N', Figure 1. If PUL is loose, the patients may have 'mixed incontinence', Figure 1, and both may be cured by MUS. Bladder emptying is facilitated by relaxation of the forward vector (arrow), Figure 1. The posterior vectors (arrows) now pull open the posterior urethral wall (broken lines, bladder), to decrease resistance to flow. Lax USLs weaken the posterior vectors which contract against them.

Causation pathway for fecal incontinence and obstructive defecation syndrome

The posterior levator plate (LP) vector, Figure 1, inserts into the posterior wall of the rectum. For



Figure 1. Relationship of symptoms and prolapse to damaged ligaments in chronic pelvic pain of unknown origin. (CPPU) almost invariably co-occurs with bladder symptoms and bowel symptoms, proportionally as indicated on the left figure. Three directional forces (arrows) contract against pubourethral (PUL) anteriorly and uterosacral (USL) ligaments posteriorly to close or open (broken lines) urethral and anal tubes. Loose ligaments may cause specific symptoms as indicated. Height of bar indicates frequency of occurrence with either PUL or USL laxity. Right upper figure: Frankenhauser T11–L2 (F) and sacral (S) S2–4 plexuses. If uterosacral ligaments (USL) are loose, these cannot be supported and fire off to cause pain. Right lower figure: ratio of individual posterior zone symptoms caused by USL defect.

EUL – external urethral ligament; ATFP – arcus tendineus fascia pelvis; CL – cardinal ligament; USL – uterosacral ligament; PB – perineal body

closure, the puborectalis muscle (PRM) contracts forward. LP/ longitudinal muscle of the anus (LMA) contracts down against USL to rotate the rectum around PRM to make the anorectal angle more acute, thus closing the rectum in a 'kinking' motion. For defecation, PRM relaxes; LP/LMA open out the anorectum (broken lines, rectum, Figure 1), to facilitate emptying by the contraction of the rectum. Lax USLs may invalidate both actions. This explains the seemingly paradoxical co-occurrence of fecal incontinence and obstructive defecation syndrome (ODS).

Pre-surgical validation of USL laxity as the cause of CPPU and OAB using 'simulated operations'

Simulated operation is a clinical assessment technique for assessing whether a particular ligament is causing a particular symptom.

'Simulated operation' speculum technique for the support of USLs

The aim is to support the lax uterosacral ligaments. The patient should always be tested with a full bladder. The bottom half of a bivalve speculum is placed very gently in the posterior vaginal fornix, and the percent diminution of urge or pain symptoms annotated. Alternatively, a tampon can be gently inserted into the posterior fornix. In patients with pelvic pain, gentle pressure on the cervix or hysterectomy scar by an examining finger generally reproduces that pain.

Using the speculum method, Wu Q et al., demonstrated the relief of suburethral tenderness and urge on mechanical support of the posterior vaginal fornix [19]. Even urodynamic detrusor overactivity (DO) can sometimes be reversed by digital support of the bladder base.

Clinical testing to confirm the USL origin of pain by using the Bornstein test [20]

The Bornstein Test is a valuable clinical and research tool for assessing whether chronic pelvic pain has its origins in the nerve plexuses of the uterosacral ligaments [20]. It challenges the hypothesis that vulvodynia pain arises from the vulva and interstitial cystitis pain from the bladder. Local anesthetic injected into the uterosacral ligaments to a depth of 1.5 cm at 4 and 8 o'clock relieved typical vulvodynia and interstitial cystitis pain within 5 minutes of injection for 20 minutes [20].

Surgical repair of damaged ligaments

Cardinal/uterosacral repair by tissue fixation system (TFS) tensioned tapes [5] applied a new surgical principle "creation of an artificial collagenous neoligament" described for the 1st time in the first iteration of the Integral Theory in 1990. This was experimentally validated on canines in 1990 and is the basis for repair of the pubourethral ligament in all midurethral sling operations: a strip of polypropylene tape is inserted along the length of the ligament to create a linear deposition of collagen which reinforces the damaged ligaments [5, 16].

CONCLUSIONS

USL tapes cure/improve chronic pelvic pain of unknown origin (CPPU), bladder and bowel dysfunctions by reinforcing the uterosacral ligament (USL) against which 3 directional forces contract. A weak pubourethral ligament or USL may invalidate these forces to cause incontinence, emptying and pain symptoms, all of which can be potentially reversed by using tapes to reinforce the damaged ligaments, as demonstrated.

CONFLICTS OF INTEREST

The authors declare no conflicts of interest.

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